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# **Social Cognition in Paediatric Traumatic Brain Injury: A Systematic Review and Meta-analysis**

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## **Abstract**

**Background:** Recent evidence suggests social cognitive deficits may be among the most profound and disabling consequences of childhood traumatic brain injury (TBI); however, it is only over the last decade that this area has received increasing research attention.

**Aim:** This study aims to systematically review all studies reporting on the effects of childhood TBI on social cognition. Meta-analytic techniques were employed to determine the magnitude of social cognitive deficits in childhood TBI.

**Methods:** Literature searches were conducted in electronic databases (Medline/PubMed, Scopus, Cochrane, EMBASE, PsycINFO and CINAHL) to retrieve relevant articles on social cognitive outcomes of paediatric TBI published from 2007 – 2019. The systematic review identified fourteen eligible studies, which examined the effect of paediatric TBI on five dimensions of social cognition, including emotion recognition/perception, theory of Mind (ToM), pragmatic language, moral reasoning, and social problem solving. Of these studies, eleven articles were included in subsequent meta-analyses, which included 482 children with TBI.

**Results:** Meta-analysis using a random-effects model revealed non-significant differences between TBI and typically developing (TD) control groups on measures of emotion perception/ recognition. In contrast, children and adolescents with TBI performed significantly worse than control groups on ToM and pragmatic language tasks, with small and moderate effect sizes, respectively (Hedge's  $g = -0.46$ ;  $-0.73$ ). Meta-regression indicated that post-injury social cognitive deficits were not moderated by child age. While the effect of time since injury was not statistically significant, poorer social cognitive outcomes are documented soon after injury.

**Conclusions:** Despite relatively intact basic social cognitive skills (i.e. emotion perception/ recognition) children and adolescents with TBI are vulnerable to deficits in higher-order aspects of social cognition, including ToM and pragmatic language. These findings underscore the importance of further research, using well-validated, standardised outcome instruments, in larger paediatric TBI samples. Furthermore, longitudinal prospective studies are needed to evaluate the respective contribution of injury and non-injury factors to individual variation in outcome and recovery of social cognition after paediatric TBI.

**Keywords:** children and adolescents, traumatic brain injury, social cognition, social neuroscience

## 1. Introduction

The Center for Disease Control and Prevention states that in the USA alone, traumatic brain injury (TBI) accounts for almost half a million emergency department (ED) visits per year in children aged 4 years and under (Faul et al., 2010). In the Australian context, a recent retrospective epidemiological study reported that, over a one-year period, TBI accounted for 1,115 visits to a major paediatric tertiary hospital in Melbourne (Crowe et al., 2009). TBI represents the most common cause of childhood disability and is associated with a range of physical and cognitive impairments. Recent evidence suggests that deficits in social functioning may be among the most profound and disabling consequences of paediatric TBI (Rosema et al., 2012). In particular, deficits in social cognition are frequently reported after moderate-severe TBI and have been linked to a range of negative outcomes including reduced quality of life, reduced participation and more frequent behavioural problems (Anderson et al., 2010; Cattalani et al., 1998; van Tol et al., 2011).

Social cognition refers to mental processes used to perceive and process social stimuli, cues, and the environment (Beauchamp & Anderson, 2010). Social cognition encompasses lower order, automatic processes such as emotion perception, as well as higher order processes that are controlled and conscious (Evans, 2008). Higher order social cognitive processes include cognitive theory of mind (ToM) – the ability to make inferences about others' beliefs and intentions (Beauchamp & Anderson, 2010); and affective ToM or cognitive empathy – the ability to infer what a person is feeling, which also requires intact empathy processing – the ability to share the emotional experience of others (Sebastian et al., 2012; Singer et al., 2009). These dimensions of social cognition form a critical component of socially skilled behaviour and play an important role in an individual's everyday social functioning (Guastella et al., 2010).

While the measurement of social cognition in children is largely based on direct child assessments, research in this field is limited by a lack of consistent definition, measurement and interpretation (Green et al., 2005). The boundaries of social cognition are unclear, as many of its processes also fall within definitions of communication, emotion and motivation (Adolphs, 2001). According to Adolphs (2001), the ability to perceive, interpret, and respond to socially relevant information involves a distributed neural system which integrates perceptual, emotional, and higher-order cognitive processes (Adolphs, 2001). However, many studies adopt poorly defined or inconsistent definitions of social cognition, which contribute to significant conceptual and methodological differences across studies. For example, some studies define eye movement as an aspect of social cognition (Frischen et al., 2007), whilst others conceptualise eye movement as a perceptual process that precedes

social cognition (Adolphs, 2001). Such ambiguity and inconsistency has impeded progress in the field and leads to difficulty drawing meaningful comparisons and conclusions from existing data (Green et al., 2008).

Studies of social cognition in children must also address other conceptual and methodological challenges, including the need to account for developmental, environmental and child-related factors that explain individual variation in social cognitive skills in typical and atypical development (e.g. childhood TBI). The Heuristic Model of Social Competence (HMSC; Figure 1) provides a useful framework for conceptualising the impact of childhood traumatic brain injury on various ‘levels’ or dimensions of social competence, including social cognition, social interactions and social adjustment (Yeates et al., 2007). It acknowledges that both insult-related factors (e.g. severity of injury, lesion location) and non-insult-related variables (e.g. socio-economic status, family functioning) may independently or interactively contribute to substantial individual variation in social cognitive outcomes among children with TBI.

There is also some evidence that developmental factors (e.g. age at injury) may moderate social cognitive outcomes following childhood TBI. For instance, previous research suggests that the poorest social cognitive outcomes are documented in association with TBI sustained before the age of 12 (Donders & Warschausky, 2007; Hanten et al., 2008). More recently, it has been argued that the relationship between age at injury and social cognitive outcome is better explained by a critical period model, rather than a simple linear association (Ryan, Catroppa, Cooper, et al., 2015). According to the critical period model (Dennis et al., 2014), social cognitive skills are most vulnerable to long-term disruption from TBI sustained during critical periods of development in late childhood and adolescence that coincide with the rapid maturation of social cognitive skills and their associated neural substrates (Blakemore, 2008). Since research on the relationship between age at injury and social cognitive outcomes remains limited and findings are somewhat inconsistent across studies, further research is needed characterise the precise nature and magnitude of this association in children with TBI.

[Figure 1 placed here]

Though there has been increased interest in social cognitive outcomes following paediatric TBI, the magnitude of social cognitive dysfunction remains poorly understood, and interpretation of existing literature is limited by lack of systematic evaluation of these outcomes and the factors that contribute to variability in social cognition after paediatric TBI. Using the HMSC model as a conceptual framework (Yeates et al., 2007), this study aims to systematically review all studies reporting on social cognition outcomes of paediatric TBI over a 12-year period (2007-2019). We also employ meta-analytic techniques to determine the magnitude of social cognitive dysfunction and factors contributing to poorer social cognition in children and adolescents with TBI.

## **2. Methods**

### **2.1 Protocol registration**

The systematic review protocol was registered with the International Prospective Register of Systematic Reviews (PROSPERO: registration ID – CRD42017055483). The meta-analytic review was conducted according to Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement guidelines (Liberati et al., 2009; Moher et al., 2009) as recommended by the EQUATOR network (Enhancing the Quality and Transparency of Health Research). This systematic review utilised the EQUATOR network resources, including recommendations from a recent guide for systematic review publication for neuropsychologists (Gates & March, 2016), and the PRISMA study flow diagram, derived from current Cochrane standards for both new and updated reviews (Stovold et al., 2014).

### **2.2 Search Strategies**

Literature searches were conducted in electronic databases (Medline/PubMed, Scopus, Cochrane, EMBASE, PsycINFO and CINAHL) to retrieve relevant articles on social cognition in paediatric TBI published from 1<sup>st</sup> January 2007 to 31<sup>st</sup> December 2019. Searches were limited to English-language publications and human participants. Searches were performed by combining [“Theory of Mind” OR “social cognition” OR “social perception” OR “social behavior” OR “perspective taking” OR “mentalizing” OR “mind reading” OR “social adjustment” OR “social interaction” OR “social skills” OR “affect recognition” OR “facial recognition” OR “empathy” OR “pragmatic language” OR “discourse language” OR “prosody” OR “humour” OR “irony”], [“Head injury” OR “brain injury” OR “TBI” OR “traumatic brain injury” OR “brain concussion” OR “contrecoup injury” OR “diffuse axonal injury” OR “closed head injury” OR “penetrating head injury” OR “post-head injury coma”], [neonate OR infant OR newborn OR preschooler OR toddler OR child OR adolescent OR teen]. Symbols like “?” and “\*” were used as truncation to maximise search effectiveness (e.g. behaviour & behavior). In addition, the reference lists of retrieved papers were manually searched to identify relevant papers.

### **2.3 Study Selection Criteria**

The following inclusion criteria were developed and applied to both search strategies and retrieved articles: (1) study must include a group of children with traumatic brain injury; (2) social cognition outcomes were listed; (3) the sample was children and adolescents [18 years old and younger at the time of injury]; study included a healthy control group or orthopaedic control group; (4) published in the English language involving human participants; (5) published in peer reviewed journals. Articles were excluded if they reported on: (1) a single case study; (2) a non-clinical outcome study (i.e. studies validating social cognition measures); (3) treatments or

interventions with no measurement of social cognition at baseline/pre-intervention ; (4) epidemiology, population-based studies, or public health initiatives; (5) review, theoretical or opinion articles (dissertations were included to overcome issues associated with file drawer effect because of the limited research available in the field); (6) studies that did not report adequate data to calculate a mean, weighted effect (authors were contacted to obtain the raw data where this was not provided in the manuscript); (7) focal brain lesions due to other neurological insults such as stroke or brain tumour; (8) studies of acquired brain injury (unless there was a separate TBI subgroup included), non-accidental head injury, inflicted head injury, shaken baby syndrome, gunshot wounds, or studies that did not specifically exclude participants with a documented medical history of a disorder which affected the central nervous system (CNS); (9) participants with intellectual difficulties or severe medical conditions that impact cognitive functioning; (10) studies with qualitative data only; and (11) studies using duplicate data.

Identified records were rated by two independent reviewers to determine eligibility for the meta-analysis. Disagreements were resolved through discussion and consensus or with third parties to reduce the likelihood of rejecting relevant reports (Edwards et al., 2002). Study authors were also contacted on at least two occasions when insufficient information was available to make a determination regarding eligibility. All identified studies were checked to ensure that their samples were independent of each other, as data included in meta-analyses must be obtained from independent samples (Lipsey & Wilson, 2001). In potentially non-independent studies, the authors of the study were contacted via email for clarification. In cases where articles were reporting on social cognitive outcomes from non-independent samples (i.e. the same social cognitive data were presented in multiple articles addressing different research questions), we extracted data from the article reporting on the largest number of participants.

## **2.4 Validity Assessment**

There is currently no gold standard assessment of methodological study quality for observational research (Sanderson et al., 2007). Thus, the current study adopted the Newcastle-Ottawa Scale (NOS) as a tool to quantify the methodological quality of included studies (Peterson et al., 2011). The NOS is an instrument that evaluates the quality of non-randomised observational studies and has been utilized in several published systematic reviews and meta-analyses, including both cohort and case-control studies (see Appendix C). The scale implemented a star system in which each study can receive up to a maximum of nine stars if all criteria have been satisfied in all three categories: selection, comparability and exposure/outcome.

Past research has suggested that the NOS possesses adequate test-retest reliability, but its inter-rater reliability is typically poor (Stang, 2010). Thus, the current study adopted NOS as a method to quantify methodological quality as a relative measure rather than an absolute measure of quality (i.e. quantifying methodological quality on a scale for comparison, instead of rating the paper as pass/ fail in quality assessment). Further examination for any additional risks was carried out to address this issue. In order to assess publication bias, funnel plots were generated for each social cognitive factor in which standard error of the intervention effect (TBI) estimate was plotted against study sample size.

## **2.5 Data Collection Process**

The following variables were extracted and coded from eligible studies: demographics of the participants, severity of TBI [i.e. Glasgow Coma Scale (GCS) score, Loss of Consciousness (LOC) and Post-traumatic Amnesia (PTA)], and any social cognition outcome measures. Participant characteristics were also extracted when available, including sample size, age at assessment, number of participants in each group, age at injury, and intelligence quotient (IQ) post-injury. In most cases, data extracted included means and standard deviations or group means of the outcome measures of social cognition, or statistics which could be used to calculate effect sizes (e.g.  $F$ ,  $t$ ). If results at multiple time points were reported, the baseline data was extracted because it commonly had the largest sample size due to attrition at follow-up. The corresponding authors were contacted to obtain required data that was not reported. Studies were excluded if required data was not obtained after at least two attempts to contact the corresponding authors.

## **2.6 Data Analyses**

The data was analysed using the Comprehensive Meta-Analysis (Version 2, 2014) statistical package (Borenstein et al., 2005). A random-effects model, as opposed to a fixed-effects model was used given the expected heterogeneity of studies.

Due to the varying statistical metrics and techniques adopted to measure social cognition, all assessment metrics were standardised for this meta-analysis by converting each metric used to a Hedge's  $g$  (and 95% CI). Hedge's  $g$  is equivalent to Cohen's  $d$ , but includes a correction for small sample sizes (Hedges, 1981). The magnitude of Hedge's  $g$  was interpreted using Cohen's conventions with small (0.2), medium (0.5) and large (0.8). Hedge's  $g$  was calculated for each social cognition domain, and represents the difference in the domain-specific social cognition outcome measure between TBI and (healthy and orthopaedic) control groups, with negative effect sizes representing poorer social cognition in the TBI group.



This current systematic review identified five domains of social cognition in the selected studies. These domains included (1) emotion perception/recognition; (2) pragmatic language; (3) ToM; (4) social problem solving; and (5) moral reasoning. Meta-analyses were conducted only for those domains of social cognition assessed in at least 3 independent studies. Small numbers of studies precluded meta-analyses for social problem solving ( $n = 1$ ) and moral reasoning ( $n = 2$ ). As such, we conducted separate meta-analyses to examine the effect of TBI on three social cognitive domains, including emotion recognition/perception, ToM, and pragmatic language. If one study reported more than one outcome measure for a single domain, a combined, average effect was computed so that each study contributed only a single effect size. This averaged effect size has a variance that moderates the non-independent samples of each outcome that are likely to be correlated.

This random-effects model assumes the effect size obtained by individual studies varies according to subject-level sampling error, as well as differences in the study design. Heterogeneity was assessed using Cochran's  $Q$ ,  $I^2$  and  $T^2$ . A systematic difference that may influence the results in an article was shown by a statistically significant  $Q$ -statistic which indicates a heterogeneous distribution of Hedge's  $g$  between studies. Heterogeneity is generally categorised as low (25%), moderate (50%) and high (75%).  $I^2$  is the proportion of observed variance that reflects differences in true effects rather than sampling error.  $T^2$  is an estimate of the between-study variance in true effects (Borenstein et al., 2005).

**2.6.1 Moderator analysis.** To determine whether effect sizes varied according to the injury severity, age at injury, time since injury and age at assessment, a subgroup analysis was conducted using a  $Q$  test of heterogeneity. Effect sizes were calculated and compared across each subgroup. Quality of data available for these variables was evaluated to determine if it was adequate for a moderator analysis. Missing data and non-normal (or highly skewed) data will result in redundant analysis, and in this case a moderator analysis was not conducted with that variable.

**2.6.2 Publication bias and outliers.** The funnel plot was visually inspected and evaluated for potential bias by creating a scatter of study effect sizes and corresponding standard errors (Sterne et al., 2011). Rosenthal's fail-safe  $N$  ( $N_{fs}$ ) was calculated to consider potential publication bias. This determines the number of unpublished studies with non-significant results which need to be included for the  $p$ -value of the observed summary effect to no longer be significant. It is important to note that this approach focuses on statistical, rather than clinical significance. Orwin's  $N_{fs}$  was then calculated to investigate the statistical significance, by determining the number of missing studies with a Hedge's  $g$  of .00 that would be needed to 'trivialise' the effect (bring the effect size down to 0.05). Publication bias was only considered non statistically significant, if  $N_{fs}$  is higher than the tolerance

level ( $5K + 10$ ) where  $K$  represents the number of included studies (Borenstein et al., 2005). However, the  $N_{fs}$  approach indicates only if publication bias is entirely responsible for the observed effect. The ‘trim and fill’ procedure was then carried out to better estimate the impact of bias. The trim and fill procedure provides an estimate of what the effect size would be if no publication bias were present, classifying bias as ‘trivial’, ‘modest’ or ‘substantial’ (Borenstein et al., 2005).

A “One study removed” method was adopted to assess the presence and influence of outliers, which tests how the summary effect would shift if each study were individually removed from the analysis. If the studies did not substantially change the overall effect size, it was retained in the analyses.

### **3. Results**

#### **3.1 Study Sample**

The process of study selection is summarized in Figure 2, consistent with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. The systematic literature search produced a total of 2581 records, 1597 of which were unique following removal of duplicates. Of these, 1472 were excluded based on title and abstract screening, and a further 110 were excluded based on full-text screening. Inter-rater reliability for the title and abstract screening was 95.6% and for full-text screening was 90.2%. Fourteen studies met all inclusion criteria and were included in the systematic review, but only eleven studies were included in the meta-analysis due to insufficient studies in two domains.

[Figure 2 placed here]

#### **3.2 Injury characteristics of the samples**

Injury-related characteristics of the samples are summarised in Table 1. Injury factors (including TBI severity) were often measured using different classification systems or in some cases, not reported. For example, TBI severity was classified using the GCS in eight studies, duration of PTA and LOC in one study, and the Mayo Classification System was used in one study. The remaining studies did not report the TBI severity classification method used. All studies recruited participants with varying levels of TBI severity; eight of these studies reported the number of participants in each TBI severity group (i.e. mild, moderate and severe), while the remaining six did not. Among the eight studies that did, five examined social cognitive outcome according to TBI severity level, and of these, only two reported the TBI severity classification system used. Measures of social cognition were highly variable across studies and further examined below.

**3.2.1 Participants.** The total sample consisted of 482 participants with a history of childhood TBI and 583 healthy control/ orthopaedic injury control participants. The mean age at assessment of all the TBI samples

across 14 studies was 14.61 years. One study involved a sample of adults who were injured during childhood (26 to 59 years old for the control group and 30 to 55 years old for the TBI group), on average 31 years post-injury (Atay et al., 2016). Mean age at injury across 10 studies (5 studies did not report mean age at injury) for the TBI group was 9.24, and mean time since injury across 10 studies (4 studies did not report the mean of time since injury) ranged from 1 month to 31 years post-TBI. TBI samples included participants with injuries ranging from mild to severe TBI, with only six studies reporting the mean GCS scores. The mean IQ of the TBI groups ranged from 88.47 to 104.97 across studies.

**3.2.2 Study Design.** Fourteen studies utilised a cross-sectional design with a separate typically developing control/ orthopaedic injury group. One study utilised a longitudinal design (Ryan et al., 2017), which included data collected at 6- and 24-months post injury. In this study, only the 6-month post injury data was extracted due to the larger number of data points at this earlier time point.

**3.2.3 Social cognition outcome measures.** It is evident that different measurements were used by authors when examining the same construct (as shown in Table 2). The validity and limitation of this will be further explored in the discussion.

[Table 1 placed here]

### **3.3 Study Quality: Risk of Bias**

The NOS overall quality rating score ranged from 7 – 9 ( $M = 8.42$ ;  $S.D. = 0.41$ ), as reported in Table 2. Overall, the quality of studies was rated as ‘high’.

[Table 2 placed here]

In terms of selection of cases and controls, all studies demonstrated evidence of controlling for the potential of representativeness and selection bias. All studies showed adequate case definition, adopting the criteria for TBI that is aligned with the American Congress of Rehabilitation Medicine (ACRM) definition of TBI. All studies recruited participants from a designated hospital setting over a clearly defined period of time. Four studies recruited orthopaedic injury controls rather than community controls. All studies also clearly stated that controls had no history of TBI.

In terms of comparability, all studies recruited healthy controls or orthopaedic injury (OI) controls who were aged-matched to the TBI group. All studies controlled for more than one potential confounding variable, including gender, parent education level, IQ at assessment, child’s adaptive and social function, socio-economic status (SES), parent’s education and ethnicity.

With respect to TBI characteristics (e.g. TBI severity), all but one study reported that this information was extracted from patients' medical records. The non-response rate to invitations to participate was higher in the TBI than the control population in two studies. With the exception of two studies that did not report the non-responder rates, all remaining studies reported comparable response rates in the TBI and control groups.

Taken together, the NOS revealed that papers included in the present meta-analysis were of high quality, suggesting minimal methodological sources of bias.

### **3.4 Emotion Perception/ Recognition Outcomes in Paediatric TBI**

Three studies included at least one measure of emotion perception/ recognition. One of these studies reported data separately for different aspects of the same measure. None of these three studies produced a significant effect size. Meta-analysis (Figure 3) revealed no significant difference in emotion perception/ recognition between the TBI and typically developing control group (Hedge's  $g = -0.14$ , 95% CI =  $-0.48 - 0.22$ ,  $z = -0.76$ ,  $p = .45$ ). Dispersion of effects was not significant ( $Q = 0.77$ ,  $df = 2$ ,  $p = .68$ ).  $I^2$  indicated that 0% of observed variance between studies was due to differences in the true effect size.  $T^2$  indicated the variance in true effect sizes was .00.

[Figure 3 placed here]

Rosenthal's  $N_{fs}$  was 0 in this case and given that this is not greater than the tolerance level ( $5K + 10$ ), there was evidence of publication bias. The trim-and-fill procedure indicated that no further studies would need to be included to make the plot symmetrical (see Figure 4). The adjusted Hedge's  $g$  remained in between CIs. Thus, the effect of publication bias on the overall effect size was probably trivial. The one study removed procedure indicated that with the removal of any one study, Hedge's  $g$  remained non-significant. Therefore, no studies were considered outliers and were retained.

[Figure 4 placed here]

### **3.5 Social Problem Solving Outcomes in Paediatric TBI**

Social problem solving was examined in only one study. This study evaluated social problem solving using a task involving eight hypothetical situations involving real-world social problems (Ganesalingam et al., 2007). Children with TBI were found to have poorer social problem solving skills, such that the TBI group were significantly more likely to select aggressive and avoidant solutions to the hypothetical problems. The TBI group was also significantly less likely to choose assertive solutions (Ganesalingam et al., 2007). Cognitive and behavioural self-regulation skills were positively correlated with assertive solutions and negatively correlated with aggressive solutions. In addition, social problem solving skills were a significant independent predictor of

social and behavioural functioning, such that aggressive and avoidant solutions were linked to worse social and behavioural adjustment. Overall, this study suggests that children with TBI are more likely to choose aggressive and avoidant solutions to social problems, and that these maladaptive responses are associated with poorer self-regulation and worse everyday social and behavioural functioning.

### **3.6 Pragmatic Language Outcomes in Paediatric TBI**

Four studies included at least one measure of pragmatic language. Two of these studies reported data separately for different subtests of the same measure. Of these four studies, three studies showed that relative to typically developing controls, children with TBI displayed significantly worse performances on well-validated measures of pragmatic language, including the Test of Language Competence – Expanded Edition (TLC-E) and Comprehensive Assessment of Spoken Language (CASL) – Pragmatic Judgement subtest. The remaining study examined group differences on The Awareness of Social Inference Test (TASIT) and found no statistically significant effect of TBI on pragmatic language outcome (McDonald et al., 2013).

Meta-analysis of these four studies (see Figure 5) revealed that overall, the TBI groups performed significantly worse than control groups on pragmatic language tasks, with a moderate effect size (Hedge's  $g = -0.73$ , 95% CI =  $-0.99 - -0.47$ ,  $z = -5.56$ ,  $p < .001$ ). Dispersion of effect sizes was significant ( $Q = 2.43$ ,  $df = 4$ ,  $p = .66$ ).  $I^2$  indicated that 0% of the observed variance between studies was due to differences in the true effect size.  $T^2$  similarly indicated that the variance in true effect sizes was .00.

[Figure 5 placed here]

Rosenthal's  $N_{fs}$  was 33, and this is not greater than the tolerance level ( $5K + 10$ ), indicating some evidence of publication bias. On the other hand, the trim-and-fill procedure indicated that no further studies would need to be included to make the plot symmetrical (see Figure 6). The adjusted Hedge's  $g$  remained in between CIs. Thus, the effect of publication bias on the overall effect size is likely trivial. The one study removed procedure indicated that with the removal of any one study, Hedge's  $g$  remained significant, and thus no studies were considered outliers and all included studies were retained.

[Figure 6 placed here]

### **3.7 ToM outcomes in Paediatric TBI**

Seven studies included at least one measure of ToM. Four of these studies included more than one measure of ToM or reported data separately for different aspects of the same measure. Of these seven studies, four studies showed a significantly poorer ToM performance in children with TBI (Genova et al., 2019; Levy &

Milgram, 2016; Robinson et al., 2014; Sirois et al., 2017). Although the remaining three studies also reported lower ToM performance in TBI groups, the differences did not reach statistical significance.

Meta-analysis of these seven studies (see Figure 7) revealed that overall, TBI groups have worse ToM when compared to control groups, with a small effect size (Hedge's  $g = -0.46$ , 95% CI =  $-0.63 - -0.28$ ,  $z = -5.18$ ,  $p < .001$ ). Dispersion of effect sizes was not significant ( $Q = 9.20$ ,  $df = 6$ ,  $p = .16$ ).  $I^2$  indicated that 34.74% observed variance between studies was due to differences in the true effect size. In contrast,  $T^2$  indicated that the variance in true effect sizes was .03.

[Figure 7 placed here]

Rosenthal's  $N_{fs}$  was 52 and given that this is larger than the tolerance level ( $5K + 10$ ), there was no significant concern of publication bias. Similarly, the trim-and-fill procedure indicated that no further studies would need to be included to make the plot symmetrical (see Figure 8). The adjusted Hedge's  $g$  remained in between CIs. Thus, the effect of publication bias on the overall effect size was probably trivial. The one study removed procedure indicated that with the removal of any one study, Hedge's  $g$  remained significant. Therefore, no studies were considered as outliers and were all retained.

[Figure 8 placed here]

### **3.8 Moral Reasoning outcomes in Paediatric TBI**

Two studies included a measure of moral reasoning (see Figure 9). Both studies reported on the same measure in two different TBI samples (Beauchamp et al., 2013; Beauchamp et al., 2019). For both studies, children with TBI demonstrated less moral reasoning maturity than the age-matched control group. Interestingly, scores on measures of intellectual functioning and empathy were associated with moral reasoning maturity (i.e. ability to justify response), but not moral decision making ability (i.e. ability to make a reasonable response). Of note, moral reasoning ability did not predict everyday behavioural and adaptive functioning (Beauchamp et al., 2013; Beauchamp et al., 2019).

### **3.9 Moderator Analysis**

To examine the effect of relevant clinical and demographic variables (mean age at assessment, mean age of injury, GCS score, and mean time since injury) on social cognition, a meta-regression was planned. Due to the limited number of studies within each social cognition domain, meta-regression was conducted on the overall sample of studies. A combined, average effect size was computed for studies that included more than one measure of social cognition. This is to ensure that each study contributed only to one effect size. The results of this meta-regression should therefore be interpreted with caution.

The measure of TBI severity varied across studies. GCS was most used but only six studies provided the mean GCS for the TBI group. While some studies did provide the mean GCS for each TBI severity group, meta-regression only allows for one mean score to be included in the analysis. As such, use of the mean GCS (including all TBI severity levels) substantially reduces variation in GCS across the included studies. The mean GCS ranged from 6 – 12 across the six studies. Due to the lack of variation in GCS score (mean GCS ranged from moderate to severe), a meta-regression was not conducted with this variable as it would not allow the authors to study the variation inherent across mild, moderate and severe TBI.

To gain some understanding of the effect of TBI severity on social cognition outcome, papers that reported on social cognition outcome according to severity group were further examined. Five studies examined severity group differences in social cognitive outcome. Two studies reported that children who sustained severe TBI performed worse on ToM tasks when compared to moderate TBI (Robinson et al., 2014; Walz et al., 2009). However, the difference between moderate and severe TBI was not observed across all three (cognitive, affective and conative) ToM tasks (Ryan et al., 2017). For studies involving moral reasoning, findings did not reveal any significant group differences between children with mild versus moderate/severe injuries (Beauchamp et al., 2013; Beauchamp et al., 2019).

There was sufficient data available to explore the effect of age-related variables on social cognition in meta-regression analyses. Table 3 shows that age at assessment, age at injury and time since injury did not moderate the relationship between TBI and social cognition in meta-regression analyses. However, although time since injury did not reach statistical significance, a small to medium effect size was observed.

[Table 3 placed here]

## **4. Discussion**

This meta-analysis reveals that while basic social cognitive abilities remain relatively intact, children and adolescents with TBI are most vulnerable to deficits in higher order aspects of social cognition, including pragmatic language and ToM. Despite increased interest in social cognitive outcomes of paediatric TBI over the last decade, advances in research has been limited by a dearth of age-appropriate, standardised assessment tools (with several authors developing their own experimental measures) and limited consensus on the specific social cognitive domains most vulnerable to the effects of TBI. In addressing substantial gaps in current knowledge of the social cognitive consequences of paediatric TBI, we aimed to systematically review all studies reporting on social cognitive outcomes in children and adolescents with TBI between 2007 and 2019. Guided by the HMSC conceptual framework (Yeates et al., 2007), we also employed meta-analytic techniques to determine the magnitude of social cognitive dysfunction and explored the contribution of age-related factors (age at injury, age at assessment and time since injury), TBI severity, and IQ to social cognitive outcomes after paediatric TBI. This systematic review identified fourteen eligible studies, which examined the effect of paediatric TBI on five domains of social cognition. Of these fourteen studies, eleven studies were included in subsequent meta-analyses.

### **4.1 Main findings**

Meta-analyses could be conducted for three of the five social cognitive domains, including emotion perception, ToM and pragmatic language. Although social problem solving and moral reasoning were evaluated in a small number of studies ( $n=1$  and  $n=2$ , respectively), there was insufficient data to conduct meta-analyses of these outcomes. Interestingly, findings revealed preferential disruption to specific domains of social cognitive ability. Specifically, we found that while paediatric TBI was associated with significantly poorer ToM and pragmatic language, the effect of TBI on emotion perception/recognition was non-significant. While these findings lend support to the vulnerability of the immature ‘social brain’ to disruption from TBI, it appears that the basic level of social cognition remains relatively preserved.

### **4.2 Results in the Context of Literature**

It has been a common assertion in the literature that children with TBI display social cognitive deficits (McDonald et al., 2014; Rosema et al., 2012). Our meta-analytic findings are broadly consistent with this claim; however, based on current evidence it appears that these deficits are limited to the domains of ToM and pragmatic language, with small-medium and medium-large effect sizes observed, respectively.

Findings in the ToM domain should be interpreted in the context of recent theoretical models of ToM, which distinguish between basic and higher order aspects of this construct that were not universally examined in



all studies. For instance, a previous study proposed the Tripartite ToM model which distinguishes between cognitive ToM (i.e. basic, false belief understanding) that undergoes rapid maturation in infancy and early childhood, and more complex, higher order aspects of conative and affective ToM which show extended developmental trajectories (Dennis et al., 2013). While affective ToM involves the understanding that facial expressions are often socially modulated to communicate emotions that individuals want others to think they feel (Dennis et al., 2013; Hein & Singer, 2010), conative ToM is concerned with the ability to understand how indirect speech involving empathy and irony are used to influence the affective or mental state of the listener (Dennis et al., 2013).

In the majority of studies included in this review and meta-analysis, ToM was typically examined as a single construct tapped using basic false belief type tasks, rather than as three separate dimensions proposed by the Tripartite model of ToM (Dennis et al., 2013). Only one study tested this model that is included in this meta-analysis (Ryan et al., 2017). In this study, the author report that while conative and affective ToM were most vulnerable to the effects of TBI, cognitive ToM is relatively spared. The differential impact of pediatric TBI on various aspects of ToM is likely explained by developmental factors, whereby skills that are better established at the time of injury (i.e. cognitive ToM) are less vulnerable than those skills that are emerging or not yet developed at the time of injury (i.e. conative and affective ToM). Given that the majority of studies included in our analyses employed measures that more closely approximate cognitive aspects of ToM, it is possible that existing research does not adequately capture the effects on more complex, higher order aspects of ToM that require further investigation.

For lower-order aspects of social cognition like emotion perception/recognition that develop relatively early in life, differences between TBI and control groups did not reach statistical significance in the current study, consistent with previous research showing that skills that are well established at the time of injury are less vulnerable to paediatric TBI (Dennis et al., 1996). Since emotion perception/recognition is acquired at a younger age, it is not surprising to see relatively preserved emotion recognition performance for children who sustained TBI in the included age range. Past studies have suggested that emotion recognition performance after TBI may vary as a function of stimulus modality (McDonald & Pearce, 1996). Although deficits in recognising emotion in both face and voice normally co-exist, they are often dissociable (Adolphs et al., 2002; Hornak et al., 1996). While the four studies included in the current meta-analysis involved both face and voice emotion recognition, the differences of modality of deficits could not be tested.

Previous evidence suggests that childhood TBI is associated with impairments on tasks involving higher-order cognitive skills and decision making (Muscara et al., 2008). The current meta-analytic findings offer broad support for this suggestion. Specifically, our meta-analytic results show that, while basic emotion recognition skills remain relatively intact following early brain insult, children with TBI display significantly poorer performance on higher-order social cognitive tasks involving both ToM and pragmatic aspects of language. Due to the small number of studies involving measures of social problem solving and moral reasoning, meta-analysis of these social cognitive domains was not possible. As such, further research is needed to assess whether the effects of paediatric TBI on higher-order aspects of social cognition generalise to these other important social abilities.

### **4.3 Potential Moderators**

Given the substantial individual variation in social outcomes among survivors of child TBI (Yeates et al., 2007), our analyses also aimed to evaluate the potential injury and non-injury related moderators of post-injury social cognition. Though there has been preliminary evidence for a dose-response relationship between injury severity and a range of social outcomes measured in the extant literature (Catroppa & Anderson, 2004; Ryan et al., 2014), meta-regression to examine the role TBI severity was not possible owing to substantial variability in study methodology. When comparing across individual studies at a descriptive level, the effect of TBI severity appeared mixed. Two studies suggested that severe TBI was associated with significantly worse social cognitive outcomes than moderate TBI (Robinson et al., 2014; Walz et al., 2009), however these group differences were non-significant in a study by (Ryan et al., 2017).

Age-related factors, including age at injury, age at assessment, and time since injury did not significantly moderate social cognitive outcomes in the current meta-analysis. Findings from several studies suggest a deleterious effect of younger age at injury on various intellectual, cognitive, academic, and behavioural outcomes (Donders & Warschausky, 2007; Hanten et al., 2008; Yeates et al., 2004). The effect of younger age at injury on outcome is most consistently documented for intellectual functioning, with evidence suggesting that children sustaining TBI prior to 7 years of age have significantly poorer outcomes than those children sustaining TBI at an older age (Anderson et al., 2000; Radcliffe et al., 1994). Although our meta-aggression analyses found that age did not moderate social cognitive outcomes, it is likely that the relationship between age and social cognitive outcomes is not explained by a simple linear association. Instead, there is recent evidence that the relationship between age at injury and social cognition outcomes is explained by a critical period model. This model suggests that for any given social cognitive domain, outcomes are likely dependent on the child's developmental stage at

the time of injury. This model would predict that higher-order social cognitive skills (e.g. complex ToM, pragmatic language) are most vulnerable to persisting, long-term disruption from TBI sustained during late childhood and early adolescence; developmental periods that coincide with rapid maturation of higher-order social cognitive skills and associated neural circuitry. Although this model receives preliminary support from previous studies of ToM and pragmatic language after paediatric TBI (Ryan et al., 2014), further studies are needed to model the likely complex relationships between age at injury and social cognitive outcomes.

Finally, although time since injury was not a significant moderator of social cognitive outcomes, this variable was associated a high coefficient of determination. Although the direction and magnitude of this effect suggests that worse social cognitive outcomes are documented soon after injury, further longitudinal studies are needed to evaluate the contribution of time since injury to outcome and recovery of social cognition in larger samples of children with TBI.

#### **4.4 Strengths of this study**

Of note, the initial search yielded 124 relevant studies; however only 11% met the review criteria. This suggests that, consistent with the emerging nature of this research domain, many existing studies in this field are characterised by significant methodological weaknesses, including poorly defined exclusion and inclusion criteria, and a notable absence of standardised assessment measures and appropriate control groups. As a result of our rigorous exclusion criteria, NOS was high and included studies controlling for potential methodological sources of bias. As such, this systematic review and meta-analysis represents a robust synthesis of high-quality evidence from this emerging research area.

#### **4.5 Study Limitations**

One of the most significant methodological limitations of research in this area is the limited number of studies available for entry into the meta-analysis. There was also substantial variability in definitions and measurement tools used to assess social cognition. Findings from some social cognitive domains (e.g. moral reasoning) are weakened by inconsistent definitions and the absence of well validated, standardised assessment tools. Specifically, several studies have relied on novel experimental measures with limited normative data, (Beauchamp et al., 2013; Beauchamp et al., 2019; Ganesalingam et al., 2007). Even where well-established measures are available, different studies adopted different measurement tools to assess the same construct. For example, pragmatic language was assessed using three different tests: the TASIT, TLC-E and CASL (Atay et al., 2016; McDonald et al., 2013; Ryan et al., 2017; Turkstra et al., 2008). This resulted in lower comparability across studies. Similarly, while many social cognitive tests purport to assess the same underlying construct (e.g.

pragmatic language), these measures often differ in task complexity and difficulty level. For instance, complex measures of pragmatic language often place greater demands on general abilities, such as working memory and cognitive flexibility, which may at least partly account for group differences in outcome.

There were also inconsistencies in the data reported across studies. Several studies reported the age at assessment but not the age at injury, which contributed to difficulties in examining recovery trajectories for social cognition. Moreover, age at assessment and age at injury covered a broad range, which precluded the examination of crucial developmental stages. The definitions of injury severity were also inconsistent across different studies. Some included a “complicated mild TBI” group, while others combined the “moderate and severe TBI” group into one category. GCS scores were also often not reported, with some studies reporting PTA/LOC scores instead, and one study adopted the Mayo Classification System. Due to the inconsistencies in reporting these data, in addition to significant amounts of missing data (mean GCS scores of each severity group), meta-regression for severity of injury was not possible.

Although a wide range of databases were searched (i.e. six in total), unpublished studies were not included in this review. Moreover, only studies published in the English language were included, which resulted in limited generalisability to other cultures. While efforts were made to contact authors by email for missing data, some studies were not included despite multiple attempts to contact these authors. Nevertheless, visual inspection of funnel plots for each social cognitive domains suggests that publication bias is unlikely.

#### **4.6 Implications and Future Direction**

Our meta-analytic findings reveal that while basic social cognitive abilities remain relatively intact, children and adolescents with TBI are most vulnerable to deficits in higher order aspects of social cognition, including ToM and pragmatic language. It is well established that impaired ToM and pragmatic language may contribute to significant everyday social difficulties, including reduced social competence, social isolation and poorer social integration (Cacioppo, 2002; Mateer & Sira, 2006; McDonald et al., 2003; McGann et al., 1997; Ownsworth & Fleming, 2005). Nevertheless, it is equally important to recognise the limitations of social cognitive tests, including issues concerning ecological validity (Hermans et al., 2019). As such, for some children, impairments in everyday social functioning may be evident, even in the absence of impairments on formal social cognitive tests. Despite the well-recognised limitations of social cognition tests, our findings underscore the importance of routine clinical screening for social cognition deficits following childhood TBI. Traditionally, social cognitive deficits have received relatively less attention in rehabilitation research than other aspects of motor and cognitive function (Bornhofen & McDonald, 2008). The results of this meta-analysis thus underscore

the importance of incorporating social cognitive assessment tools into routine clinical practice, as well as a critical need to develop theory-driven, evidence based treatments for social cognitive impairment after childhood TBI.

Moreover, due to the small number of studies reporting neuroimaging findings, we were unable to evaluate the potential contribution lesion characteristics (e.g. lesion location, lesion size and overall lesion burden) as moderators of post-injury social cognitive outcomes. Preliminary findings have documented worse social cognitive outcomes in association with larger and more diffuse haemorrhagic lesions (Ryan, Catroppa, Beare, et al., 2015; Ryan, Catroppa, Cooper, et al., 2015); however, further studies are needed to replicate these findings in larger samples of children with TBI. Additional research is needed to explore the potential contribution of non-injury variables to outcome and recovery of social cognition after paediatric TBI. Family environment and social context are well-established influences for social development (Ackerman & Brown, 2006; Guralnick et al., 2003; Masten et al., 1999). In the context of child TBI, socioeconomic status, family dysfunction, and permissive parenting are known to predict poor social competence (Chapman et al., 2010; Yeates et al., 2004). Since studies included in the current review did not report data on non-injury variables, further longitudinal research is needed to understand the role of the proximal and distal family environment in moderating social cognitive outcomes after TBI. Such data is likely to inform the design and implementation of context-sensitive, family based interventions to improve social cognitive outcomes.

Finally, given the dearth of longitudinal research in this area, further studies are needed to evaluate the effect of TBI severity on longitudinal recovery trajectories, particularly in children with severe TBI. Due to small sample sizes, outcomes of children with severe TBI are often not analysed separately from other TBI severity groups, making findings difficult to generalise and interpret. Large prospective studies using common data elements from multiple sites are likely to significantly advance our understanding of social cognition after childhood TBI and address some of the identified challenges.

#### **4.7 Conclusion**

The findings of the current study synthesise a somewhat disparate body of literature of social cognition in paediatric TBI. Overall, our meta-analytic findings reveal that while basic social cognitive abilities remain relatively intact, children and adolescents with TBI are most vulnerable to deficits in higher order aspects of social cognition, including ToM and pragmatic language. Further studies employing longitudinal prospective designs with larger samples of more severely injured children are needed to better delineate injury-and non-injury-related

factors influencing outcome and recovery of social cognition after child TBI. Such efforts will be beneficial for informing the design and implementation of theory driven, evidence-based treatments for social cognitive impairment after child TBI.

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